Pelvic Osteoid Osteoma in a Skeletally Mature Female

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Abstract

Osteoid osteoma is the most common bone-producing tumor that typically presents with "throbbing night pain" and that improves dramatically with use of low-dose salicylates. Few cases of pelvic osteoid osteoma have been reported, and most have involved patients younger than age 30. Surgical excision classically has been the treatment of choice, but, recently, less invasive modalities, including radiofrequency ablation, have begun to supplant surgical management of osteoid osteoma, resulting in a decrease in the need for definitive surgical diagnosis and treatment. We present a rare case of osteoid osteoma in the pelvis of a woman older than age 30.

A 37-year-old woman presented to her primary care physician after 5 months of new-onset right hip pain. She initially attributed the pain to a new exercise regimen. When the pain failed to improve after she discontinued exercising, she sought a medical evaluation. The patient provided written informed consent for print and electronic publication of this case report.

The patient localized the pain deep into the midportion of the inguinal canal, just lateral to the femoral vessels, and characterized it as a constant dull ache. Any motion, including vibrations from a car, or pressure, including sleeping on her right side, exacerbated the pain. She had trialed an anti-inflammatory medication, but it provided only temporary relief.

On physical examination, the patient was 5 ft 4 in tall and weighed 120 lb. The inguinal canal was benign with no swelling, palpable mass, or lymphadenopathy. The right groin pain was provoked with hip extension and with resisted hip abduction. The right hip had full range of motion in flexion, abduction, and internal and external rotation. There were no flexion contractures. Compression of the pelvis did not reproduce the pain. Both legs were neurovascularly intact distally with intact sensation to light touch and palpable dorsalis pedis pulses. The patient was able to stand on each leg and walk without exacerbation of the groin pain.

The primary care physician conducted a thorough workup, which included pelvic ultrasound and complete genitourinary system evaluation with a consulting gynecologist, as well as colonoscopy and complete gastric system evaluation with a consulting gastroenterologist. Results of all these evaluations were negative. Subsequently, an anteroposterior radiograph of the pelvis was obtained; it showed sclerosis of the right iliac

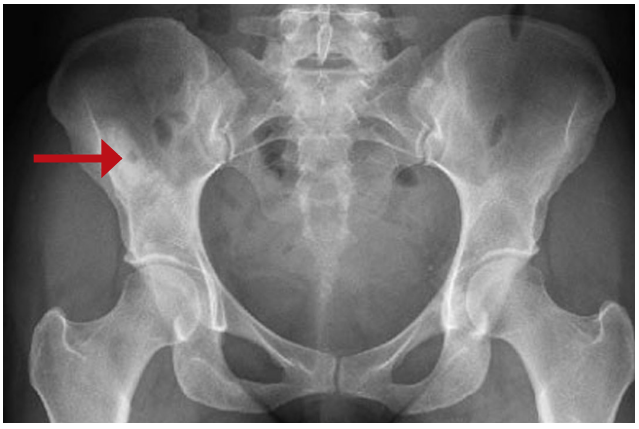

Figure 1. Anteroposterior radiograph of pelvis shows radiodense lesion of right iliac wing with radiolucent nidus (arrow).

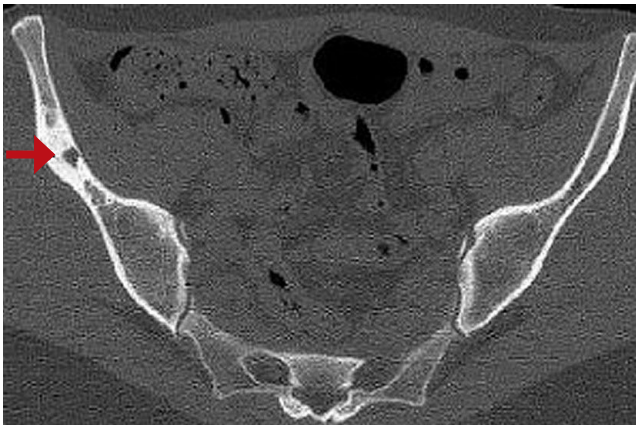

Figure 2. Computed tomography scan of pelvis shows sclerotic lesion of right ilium with radiolucent nidus (arrow) less than 2 cm in diameter.

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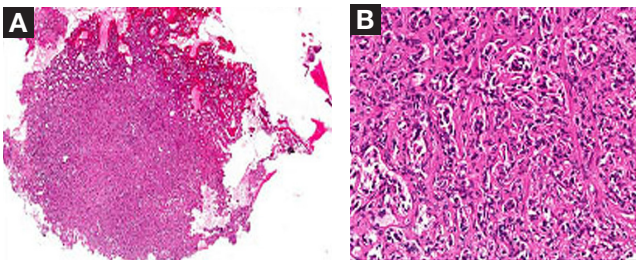

Figure 3. **(A)** Low-power (4×) view of histopathology shows well-circumscribed nidus. **(B)** High-power (10×) view of histopathology shows anastomizing bone trabeculae lined by plump osteoblast with scattered osteoclast-like giant cells, as well as vascular stroma consistent with osteoid osteoma.

wing (Figure 1). At that time, the differential diagnosis included intracortical bone abscess, chronic sclerosing osteomyelitis, bone island, tuberculosis osteomyelitis, osteoblastoma, Ewing sarcoma, osteochondritis, eosinophilic granuloma, and bony metastasis.

Advanced imaging of the pelvis was obtained. Computed tomography (CT) showed a radiodense focus within the right ilium with a round central radiolucency with no evidence of cortical disruption, periostitis, or osteolysis (Figure 2). Magnetic resonance imaging (MRI) showed no enhancement with gadolinium, no soft-tissue mass, and minimal edema around the ilium lesion on T_2-weighted images. At this point, the most likely diagnoses were osteoid osteoma, eosinophilic granuloma, and, possibly, infection, and it was determined that a biopsy was warranted. CT-guided biopsy was attempted but resulted in a nondiagnostic sample. Therefore, an open biopsy was recommended.

The patient underwent open biopsy of the right ilium lesion. An osteolytic region easily was identified deep to the psoas muscle, and curettage was used to excise the entire lesion. Histopathology revealed a well-circumscribed nidus (Figure 3A) composed of anastomizing bone trabeculae lined by plump osteoblast with scattered osteoclast-like giant cells as well as vascular stroma, consistent with osteoid osteoma (Figure 3B). By 3 months after curettage, the patient had returned to work with complete resolution of her activity-related pain.

Discussion

Osteoid osteoma is the most common bone-producing (ie, osteoblastic) tumor. First described by Jaffe in 1935, it is characterized on gross pathology and radiographic imaging as having an osteoid nidus with a surrounding region of bone sclerosis.[1,2] It typically presents with "throbbing night pain," which dramatically improves with use of low-dose salicylates.[3] Osteoid osteomas account for 10% to 12% of all bone lesions and most commonly affect men between 10 and 25 years of age.[3-5] They are relatively rare in patients older than age 30 (only 13% incidence).[3] There is a slight preponderance in men compared with women (reported ratios between 2:1 and 3:1).[3,6] More than half of all osteoid osteoma cases occur

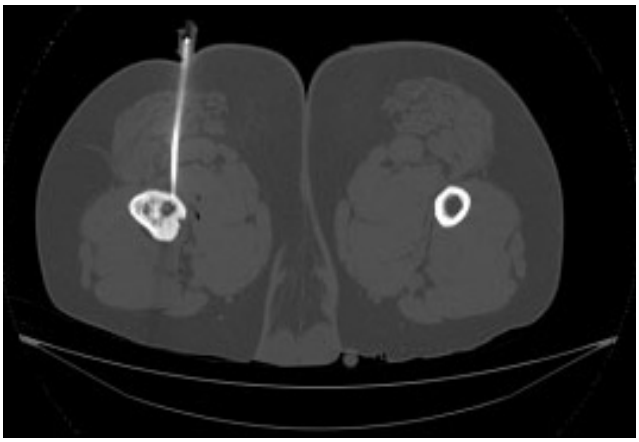

Figure 4. Computed tomography scan of pelvis shows radiofrequency ablation of osteoid osteoma in proximal femur.

in the diaphysis of the proximal femur.[4] Another 10% to 25% of cases occur in the spine.[4,7] Thirteen percent of all cases are intra-articular, and these normally involve the hip, yet only 1.2% to 2.7% of all osteoid osteoma cases occur in the pelvis.[4-8] Osteoid osteoma is a benign bone tumor with limited growth potential and a very low rate of malignant transformation. The diagnosis of osteoid osteoma is often elusive because of variability in tumor location or lack of classic radiographic findings.[2,3,9]

Few cases of pelvic osteoid osteoma have been reported, and most have involved patients younger than age 30. A 21-year-old man with right hip pain, exacerbated by activity and significantly relieved with use of nonsteroidal anti-inflammatory drugs (NSAIDs), was given an initial diagnosis of sacroiliitis.[6] However, normal inflammatory markers prompted ordering a CT scan, which showed a subcortical radiolucent region in the ilium, and osteoid osteoma was finally diagnosed. In the case of a 16-year-old girl with a 9-month history of right groin pain, pelvic radiograph showed acetabular sclerosis, CT showed a radiolucent region in the acetabular fossa, and histology showed the pathognomonic osteoid nidus of osteoid osteoma.[10] In a 13-year-old boy with sacrococcygeal pain that failed to resolve after 1 month, that worsened at night, and that was relieved with NSAID use, pelvic radiographs were normal, and pelvic CT showed a 15-mm hypodensity.[11] Osteoid osteoma was later diagnosed with a surgical pathology specimen.

The clinical manifestation of osteoid osteoma, however, may vary quite a bit. Patients with osteoid osteoma typically present with intense localized pain (median duration, 14 months).[3,7] Pelvic osteoid osteoma may present with symptoms that mimic numerous orthopedic and rheumatologic conditions, including inflammatory arthritis, degenerative arthritis, neoplasm, and infection.[12] Nocturnal pain is the most common symptom, occurring in 80% of patients with extra-articular osteoid osteoma.[13] Salicylates (ie, aspirin and NSAIDs) often reduce pain significantly, but this effect is nonspecific, as most patients lack signs or symptoms of a sys-

temic inflammatory process, and inflammatory markers are typically within normal limits.[3,6]

Delay in the diagnosis of osteoid osteoma is not uncommon. The pathognomonic osteoid nidus is not always seen on conventional radiographic imaging, and pain may mimic many other conditions depending on tumor location.[14] Histologically, the lesion is characterized by active osteoblasts (ie, osteoblastic rimming) and connective tissue with a nidus of osteoid.[3] The lesion is surrounded by reactive bone resulting from the high pressure exerted by the tumor on the surrounding normal bone.[7] On standard radiograph, this lesion may appear as a radiolucent nidus, typically less than 1 cm in size, with a ring of sclerotic bone.[15] Osteoblastoma should be suspected in cases with a nidus larger than 2 cm.[7] Other imaging modalities, including isotope bone scan and CT imaging, may be used to diagnose osteoid osteoma. A "double-density sign" highlights the characteristic nidus on isotope bone scan.[16] However, a radiolucent nidus is best identified with a thin-cut (1-mm) CT scan. CT allows for accurate tumor sizing and can facilitate localization for percutaneous treatment.[14] Although MRI lacks the sensitivity to reliably detect the nidus, it is highly sensitive for the reaction of the surrounding tissues and detection of intramedullary lesions.[14]

Pain relief is the main goal of treatment. The pain caused by osteoid osteoma is mediated by nerve endings within the tumor and the high level of prostaglandins within the nidus.[17] Surgical excision classically has been the treatment of choice, as the symptoms of osteoid osteoma typically resolve only after a prolonged period. Recently, less invasive modalities, including CT-guided excision, arthroscopic excision, cryoablation, and thermoablation with radiofrequency or laser, have begun to supplant surgical management of osteoid osteoma, resulting in a decrease in the need for definitive surgical diagnosis and treatment (Figure 4).[7,14] Radiofrequency ablation and other nonsurgical modalities induce irreversible necrosis measuring approximately 10 mm from the probe tip and should be avoided in lesions larger than 1 cm, in nonspherical lesions, in lesions less than 3 cm from the skin, and in lesions 1.5 cm from a neurovascular structure (larger lesions can be addressed with more advanced techniques, including needle repositioning and multiple overlapping radiofrequency ablation).[7,18,19] In addition, radiofrequency ablation and other percutaneous treatments use a small-caliber needle, making histologic confirmation of the diagnosis a low-yield effort.[20] Medical management (ie, aspirin or NSAID use) of unconfirmed osteoid osteoma has been reported. However, this treatment is prolonged and susceptible to drug sensitivity as well as medication-induced adverse effects.[21]

In summary, our patient's case not only represents a rare presentation of pelvic osteoid osteoma, in a woman older than age 30, but serves as a reminder of the educational and correlative power of pathologic diagnosis and surgical treatment when the current trend has been toward less invasive modalities.

Authors' Disclosure Statement

The authors report no actual or potential conflict of interest in relation to this article.

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This paper will be judged for the Resident Writer's Award.